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Case Report

Bochdalek hernia presenting with initial local fat infiltration of the thoracic cavity in a leukemic child

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ABSTRACT

Local fat infiltration of the thoracic cavity is a rare initial presentation of Bochdalek hernia. We report a case of Bochdalek hernia in a child with leukemia that demonstrated initial local fat infiltration of the thoracic cavity on computed tomography scan and progressed to an obvious diaphragmatic hernia on subsequent follow-up. We suggest that initial local fat infiltration of the thoracic cavity on computed tomography scan may indicate a potential diaphragmatic hernia.

Introduction

Different types of diaphragmatic hernias include esophageal hiatus hernia, Bochdalek hernia, and Morgagni hernia [1]. Although a decreasing prevalence during the period 1980-2006 was reported [2], congenital diaphragmatic hernias remain a poorly understood abnormality with a high mortality rate that cannot always be effectively managed. Although laparoscopic surgery may be used for all types of diaphragmatic hernias [1], precise diagnosis directly influences further therapeutic considerations, for example, laparoscope or observation.

In cases of Bochdalek hernia, local fat infiltration of the thoracic cavity is rare. We reported a 10-year-old male patient admitted to our institution with a chief complaint of fever, who was subsequently confirmed to have acute lymphoblastic leukemia by bone marrow aspiration and biopsy. Moreover, chest computed tomography (CT) revealed a local fat infiltration in the left-side thoracic cavity with no definite opening of the diaphragm noted. During follow-up, this patient developed a sudden abdominal pain after emesis, and chest radiograph and CT scan confirmed bowel herniation through the diaphragmatic defect. We suggest that local fat infiltration of the thoracic cavity should raise concern for possible subsequent diaphragmatic hernia in some cases.

Case report

A 10-year-old boy was admitted to our institution, with a chief complaint of fever and cough. His medical history did not show any other abnormal findings; nor did initial clinical examination: weight 38.5 kg, heart rate 90 beats/min, body

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temperature 36.9°C, and respiratory rate 26 beats/min. Also, there were no evidence of anemia, abnormal heart murmurs, rales, or other abnormal respiratory sounds. No signs of abdominal pain, peritoneal irritation, or palpable abdominal masses.

Sonography showed the maximum oblique diameter of the right hepatic lobe as 13.8 cm and spleen thickness as 4.2 cm; both results suggested a finding of hepatosplenomegaly. Chest CT scan demonstrated patchy density of the left lower lobe in lung window, which was diagnosed as focal infection (Fig. 1A). Viewed with mediastinal settings, regional low density was noted with a median CT value of −90 Hounsfield units in the left thoracic cavity, indicating local fat infiltration, but no explicit diaphragmatic opening was depicted (Fig. 1B,C, and D).

Blood chemistry analysis showed a prominent elevated white blood cell count (63.92×10^9/L) and erythrocyte sedimentation rate (46 mm/H). Other results of his blood test were not remarkable: red blood cell count, hemoglobin, platelet, prothrombin time, immunoglobulin and complement, T-SPOT, T lymphocyte, B lymphocyte, helper T lymphocytes, cytotoxic T lymphocytes, and NK cell. Bone marrow examinations, flow cytometry confirmed the diagnosis of acute lymphoblastic leukemia (B subtype, high risk).

The patient was treated with several cycles of chemotherapy. At the end of the 10th cycle of chemotherapy, 20 months after initial diagnosis, the patient developed persistent abdominal pain after sudden vomiting of gastric contents. Physical examination showed anemia, left upper abdominal pain, and borborygmus 2-3 times per minute. Chest radiograph was suspicious for left sided diaphragmatic hernia (Fig. 2A). CT scan with multiplanar reformations (Fig. 2B-D) confirmed that the left colon had herniated into the left thoracic cavity. Although surgical treatment was suggested, the parents declined.

When the initial chest CT scan was retrospectively reviewed and analyzed, there was no evidence of a definite diaphragm opening. We believe that the fat may have originated from the abdominal cavity and herniated into the thoracic cavity through a tiny opening of the diaphragm. The opening may be undetectable by CT scan and thus may be missed by radiologists; the tiny opening may subsequently

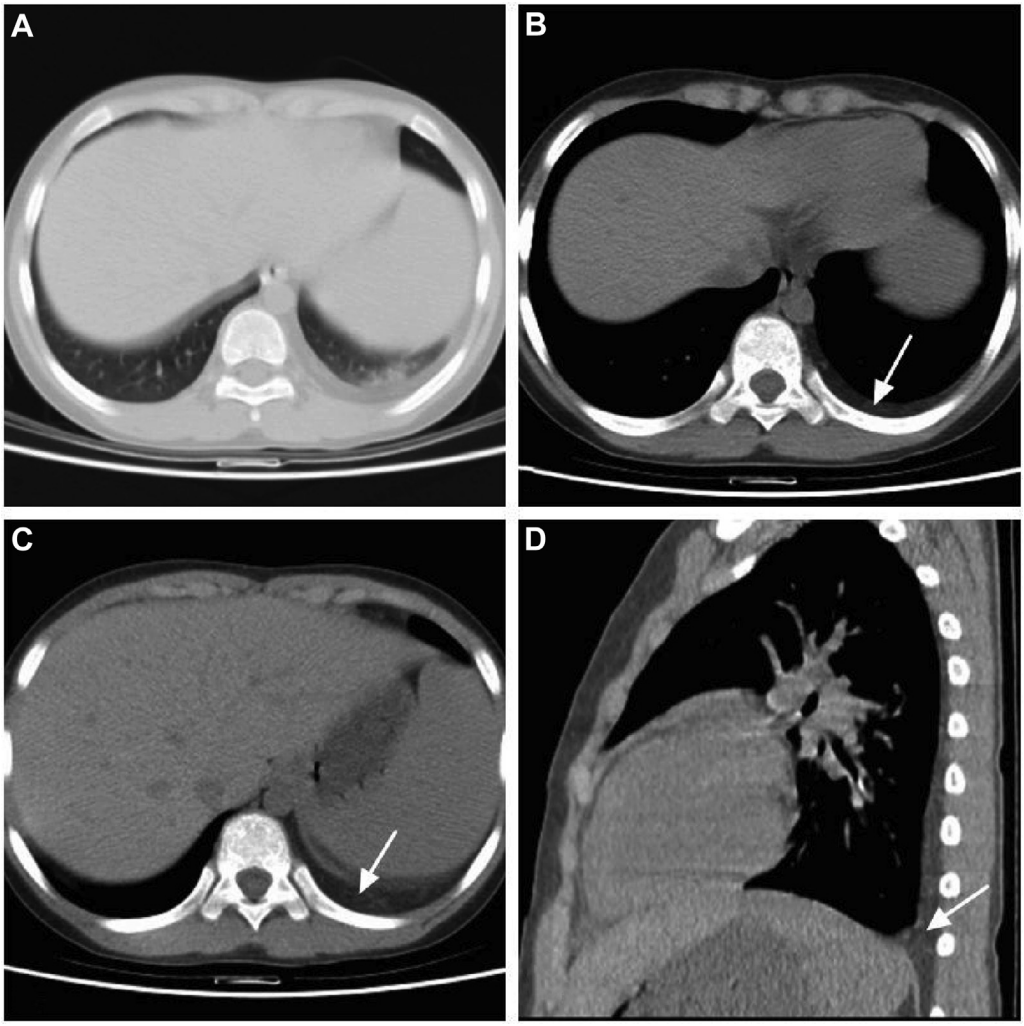

Fig. 1 – (A) Patchy density of the left lower lobe diagnosed as focal infection. (B-D) The soft tissue (white arrows) posterior to the diaphragm with average computed tomography (CT) value between −61 Hu and −91 Hu, consistent with fat tissue, diagnosed as local fat infiltration of the left thoracic cavity.

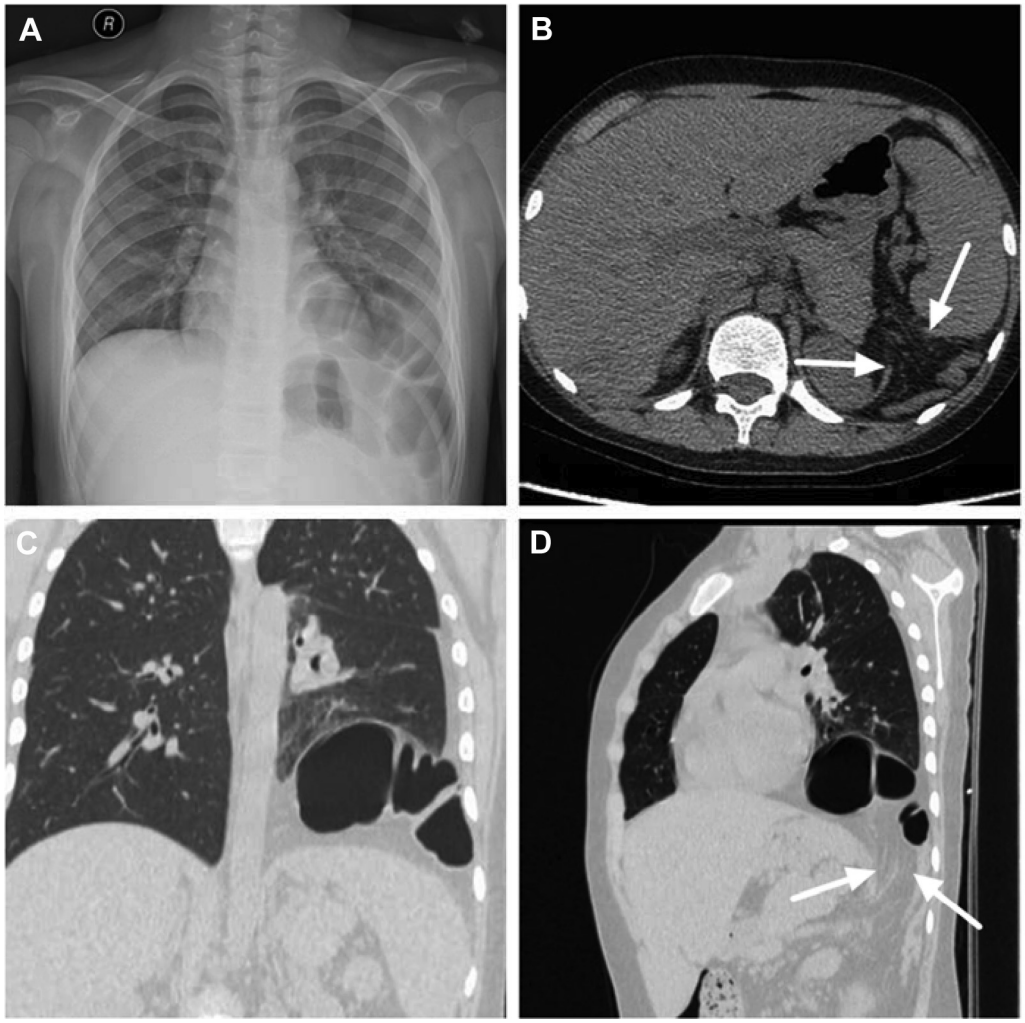

Fig. 2 – (A) The same patient 20 months postchemotherapy, x-ray showed elevation of the left diaphragm, suspicious of bowel herniation into the thoracic cavity. (B-D) CT scan demonstrated the left-sited diaphragmatic hernia with left colon herniated into the left thoracic cavity, with a clear diaphragm opening (white arrows).

progress to a large diaphragmatic hernia when abdomen pressure increases. The appearance of fat in the thoracic cavity may therefore be a diagnostic clue of potential diaphragm hernia.

Discussion

We report a rare case of Bochdalek hernia developed after a long lag time, in which the initial imaging showed local fat infiltration of thoracic cavity. This may indicate a new diagnostic clue of potential Bochdalek hernia.

We believe that this case should be classified as Bochdalek hernia, which is defined as congenital diaphragmatic defects resulting from the failure of posterolateral diaphragmatic foramina to fuse *in utero* [3]. Bochdalek hernia was first described by Vincent Alexander Bochdalek, in 1848, with a prevalence of 1 per 2500 live births [4]. Asymptomatic Bochdalek hernia is quite common among adults, with an incidence of 12.7% and age range from 36 to 86 years [5]. Left-sided Bochdalek hernia is reported by approximately 90% of cases and bilateral Bochdalek hernias are rare [6]. Most of the Bochdalek hernias are asymptomatic and thus further treatments are unnecessary. However, symptomatic patients would suffer abdomen symptoms ranging from nonspecific bloating and indigestion to the more severe complaint of intestinal obstruction [7]. Some of such patients may even suffer syncope [8] in emergent situations.

The diagnosis of diaphragm hernias has been highly associated with the development of imaging techniques. CT scan, magnetic resonance imaging, sometimes ultrasound are used to detect the diaphragm hernias and to differentiate from other diseases [9]. On imaging, Bochdalek hernia presents as a soft tissue or fatty mass abutting the surface of the posteromedial aspect of either hemidiaphragm, in continuity with subdiaphtagmatic structures through a diaphragmatic defect, presenting as a discontinuity line of the diaphragm. Usually, CT scan alone can make the diagnosis. In this case, coronal and multiplanar reformations provided additional information to better identify the diaphragmatic stripe. 3D imaging has been proven useful for

stereographic perception of the hernia [10], and could differentiate Bochdalek hernia from diaphragmatic and peridiaphragmatic masses neoplasm [11]. In addition, MRI in T1 is highly valuable to evaluate fat-containing chest lesions [12]. MRI could clearly demonstrate the mass composed of omental fat herniating into the thorax through the diaphragmatic hiatus and is considered to be a useful noninvasive modality in the evaluation of diaphragmatic hernia [13].

In this case, imaging manifestation showed a local fat infiltration of the left thoracic cavity, which was easily neglected. On retrospective analysis, this may be due to a small opening in the diaphragm, which is hard to detect by CT scan or other present imaging techniques. This is in accordance with the fact that most small diaphragm hernia would remain asymptomatic [5,11], and further intervention is not required. However, this case showed a progressive characteristic, which took a long lag time. With the expansion of the foramina, or increased intra-abdominal pressure, intestine and other abdomen organs may be herniated to the thoracic cavity, presenting a sudden and persistent tense of abdominal pain, which may cause enteric necrosis if not managed timely, until then, direct sign and indirect sign on CT scan confirmed the diagnosis.

It has been reported that Bochdalek hernias are associated with some other abnormalities, such as congenital heart defect and chromosomal abnormality [14]. However, to our acknowledge, no correlation of Bochdalek hernia and acute lymphoblastic leukemia were reported. Further analysis of this area may be needed.

Of treatments, laparoscopic surgery is a potential choice. Also, avoiding intense coughing and vomiting would benefit these kinds of patient. However, the patient's parents gave up the surgery advice.

In conclusion, a rare case was presented with radiography and CT scan, suggesting that local fat infiltration of the thoracic cavity may be a potential diagnostic clue of Bochdalek hernia.

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